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Fibrosing mediastinitis as a rare sequel of iatrogenic rupture of bronchogenic cyst — a case report

Yu-Jen Cheng [a,*], Kun-Chou Hsieh [a], Jau-Chung Hwang [b]

[a] *Division of Thoracic Surgery, Department of Surgery, E-Da Hospital/I-Shou University, 1 E-Da Road, Jiau-shu Tsuen, Yan-chau Shiang, Kaohsiung County 824, Taiwan*
[b] *Division of Anatomical Pathology, Department of Pathology and Laboratory Medicine, E-Da Hospital/I-Shou University, 1 E-Da Road, Jiau-shu Tsuen, Yan-chau Shiang, Kaohsiung County 824, Taiwan*

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Abstract Fibrosing mediastinitis is a chronic disease process with a spectrum of etiology. We report a 51-year-old female who underwent incision and drainage procedure in the neck for deep neck and mediastinal abscess. Five years later she developed fibrosing mediastinitis. This lesion infiltrated from neck base into the upper mediastinum with tracheal compression and vessel encasement. She had resection of the lesion which proved to be a ruptured bronchogenic cyst with chronic inflammation. This rare case illustrates the importance of including inflammatory bronchogenic cyst in the etiology of deep neck abscess formation. And we further find a ruptured bronchogenic cyst with chronic inflammation as an etiology of fibrosing mediastinitis.

Introduction

Fibrosing mediastinitis is an uncommon benign disorder characterized by proliferation of dense fibrous tissue within the mediastinum.[1] Though the precise cause and pathogenesis of fibrosing mediastinitis in most cases is unknown or idiopathic,[2–4] there were some reported cases with etiology identified.[1,5,6] While heterogenous in etiology there is no reported case with a ruptured bronchogenic cyst as the etiology of fibrosing mediastinitis.

Case report

A 51-year-old female, with left renal clear cell carcinoma under control, had deep neck abscess in October 2001 (Fig. 1), and underwent abscess drainage with antibiotics treatment. Recovery went smoothly until September 2006 when she developed chest pain and severe cough. Upon admission, a chest film showed an abnormal right paratracheal shadow with tracheal deviation to left side. Chest computed tomography (CT) showed diffuse mass lesion infiltrating from neck base to upper mediastinum. The right carotic and subclavian arteries were encased, and the right jugular vein and trachea were deviated outwards (Fig. 2A). The fluorodeoxyglucose positron emission tomography (FDG-PET) had an increased uptake in the lesion area. The white blood cell (WBC) count was within normal limit with only mild elevation of c-reactive protein (CRP), 16 mg/L (normal <6 mg/L). Under the impression of malignancy she underwent mediastinoscopy for tissue biopsy, which found only fibrosis tissue. To release the vessel encasement and tracheal compression the patient then underwent debulking resection through partial sternotomy. The recovery was uneventful. The final pathology reported

* Corresponding author. Tel.: +886 7 615 0011x5301.
E-mail address: yujen.cheng@msa.hinet.net (Y.-J. Cheng).

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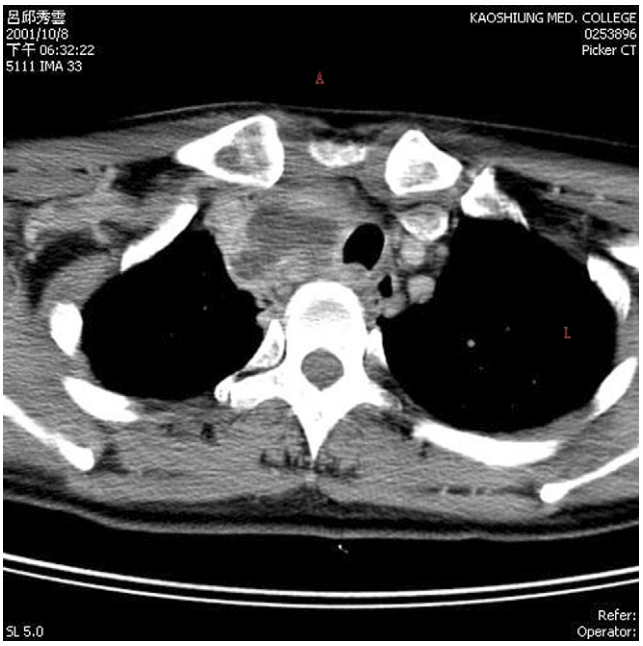

Figure 1 The deep neck abscess extending to the mediastinum was obviously viewed from the chest computed tomography.

a ruptured bronchogenic cyst with some inflammatory change (Fig. 2B).

Discussion

This is the first case of fibrosing mediastinitis, in the English literature, resulting from an unrecognized bronchogenic cyst in the upper mediastinum, when it became infected and subsequently ruptured into the adjacent soft tissue iatrogenically. At first this bronchogenic cyst was treated as a deep neck abscess extending into the upper mediastinum. The simple drainage could not completely eradicate it, which then caused a sub-clinical chronic inflammation environment in the upper mediastinum. Such environment is thought to be the inducer to form the mediastinal fibrosis.

There are two types of fibrosing mediastinitis.[2] The first type, focal mediastinal or hilar fibrosis, is caused by a fibroinflammatory response to Histoplasma capsulatum antigens. The second type, diffuse multicompartmental fibrosis, occurs idiopathically and is related to other fibroinflammatory lesions. The ever reported etiologies in the second type include aspergillosis, mucormycosis, blastomycosis, cryptococcosis, Behçet disease, rheumatic fever, radiation therapy, trauma, Hodgkin disease, drug therapy with methysergide maleate, idiopathic retroperitoneal fibrosis, sclerosing cholangitis, Riedel thyroiditis, and pseudotumor of the orbit.[1–6] In this case we further find a ruptured bronchogenic cyst with chronic inflammation as an etiology. Theoretically, when we had the Incision and drainage in 2001, this probably led to the rupture of the brochogenic cyst.

FDG-PET is a tool to evaluate the molecular activity of the lesion in contrast to the volume estimate in the CT scan. The role of FDG-PET in assessment of mediastinal pathology is controversial. However FDG-PET has the potential in the differential diagnosis between benign and malignant lesions in the thymus and lymph nodes, including thymoma,

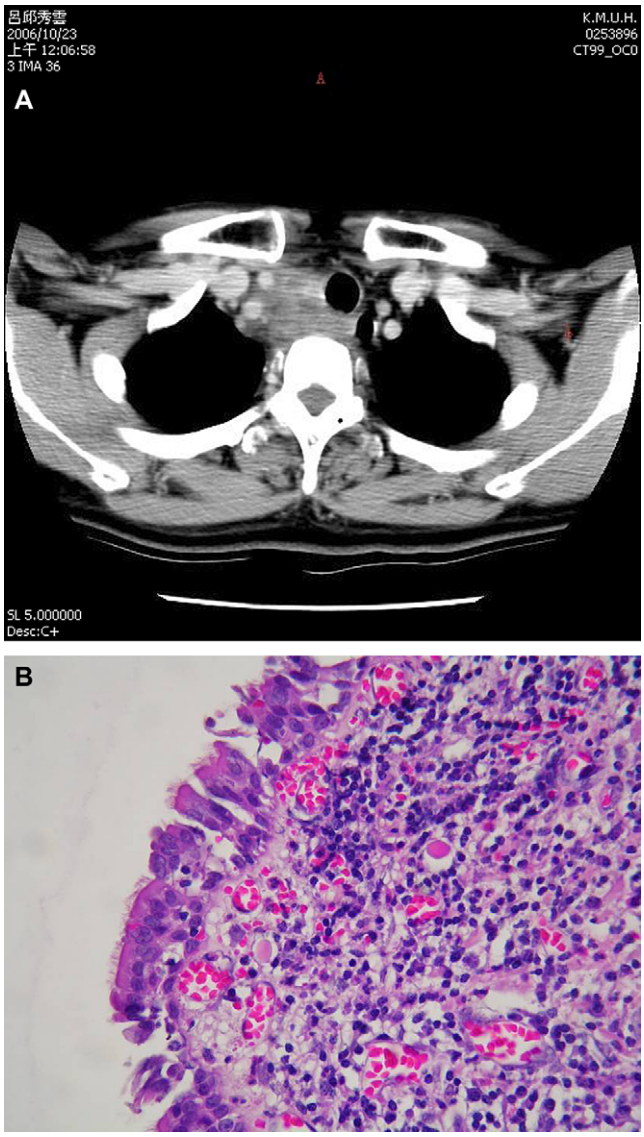

Figure 2 A. Chest computed tomography revealed the mass lesion encasing the mediastinal vessels and compressing the trachea. B. The pathology of the resected tissue showed the presence of bronchial epithelium, bronchial seromucous glands in focal area, granulation tissue formation, and some inflammation change. (Hematoxylin and eosin stain).

metastatic lymph nodes from the lung cancer and germ cell tumors.[7] Though there is a report of increased uptake in a case of mediastinal histoplasmosis,[8] which is a chronic inflammation, pre-operatively the increased uptake in this case is thought be in a malignant nature. And the moderate elevation of the CRP value further bothers us in the differential diagnosis between inflammation and malignancy, with the latter being favored. Actually the sub-clinical chronic inflammation does have such CRP value, which should be kept in mind.

References

1. Loyd JE, Tillman BF, Atkinson JB, Des Prez RM. Mediastinal fibrosis complicating histoplasmosis. *Medicine (Baltimore)* 1988;**67**: 295—310.

2. Rossi SE, McAdams HP, Rosado-de-Christenson ML, Franks TJ, Galvin JR. Fibrosing mediastinitis. *Radiographics* 2001;**21**: 737—57.
3. Dehner LP, Coffin CM. Idiopathic fibrosclerotic disorders and other inflammatory pseudotumors. *Semin Diagn Pathol* 1998; **15**:161—73.
4. Graal MB, Lustermans FA. A patient with combined mediastinal, mesenteric and retroperitoneal fibrosis. *Neth J Med* 1994;**44**: 214—9.
5. Lagerstrom CF, Mitchell HG, Graham BS, Hammon JW. Chronic fibrosing mediastinitis and superior vena caval obstruction from blastomycosis. *Ann Thorac Surg* 1992;**54**: 764—5.
6. Mole TM, Glover J, Sheppard MN. Sclerosing mediastinitis: a report on 18 cases. *Thorax* 1995;**50**:280—3.
7. Bryant AS, Cerfolio RJ, Klemm KM, Ojha B. Maximum standard uptake value of mediastinal lymph nodes on integrated FDG-PET-CT predicts pathology in patients with non-small cell lung cancer. *Ann Thorac Surg* 2006;**82**:417.
8. Mackie GC, Pohlen JM. Mediastinal histoplasmosis: F-18 FDG PET and CT findings simulating malignant disease. *Clin Nucl Med* 2005;**30**:633—5.